RESEARCH ARTICLE

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Should Children With Sub-Threshold ADHD Predominantly Inattentive Subtype (ADHD-I) Symptoms Be Treated With Sensory Integration Therapy? A Case-Control Study ABSTRACT

Objective: Sensory integration therapy is one of the promising preventive therapy options for behavioral and developmental disorders. Hypothesizing a degree of parallelism, this study provides an insight into the effectiveness of the sensory integration therapy potencies for school-aged children with subthreshold ADHD predominantly inattentive subtype.

Methods: The study was a single-arm clinical trial and 20 patients aged 7–10 years with subthreshold ADHD predominantly inattentive subtype, were included. The sensory integration intervention was prepared in accordance with sensory modulation principles and intervention strategies and lasted 12 weeks with two sessions per week. The effectiveness was assessed using the Conner's teacher/parent scales, the Clinical Global Impression scale, the Canadian Sensory integration Performance Measure and the Sensory Profile.

Results: The rate of patients with typical or better performance in auditory processing domain of the Sensory Profile were found significantly increased after sensory integration therapy; 9 patients (45%) before and 15 patients after (75%) (p=0.031). The rates of participants with typical or better performance in inattention–distractibility factor score of the Sensory Profile were found significantly increased after sensory integration therapy; 6 before (30%) and 16 after (80%) (p=0.006).

Conclusions: Sensory integration therapy focuses on supporting persons with varied disability terms to engage in daily life activities that they find significant and purposeful. Difficulties experienced by individuals with subthreshold attention-deficit hyperactivity disorder are addressed in this study and aspects of daily life are explored while swiping through different sensory modalities. Impaired auditory processing improvable through sensory integration therapy was observed in these children.

Keywords: Attention Deficit Hyperactivity Disorder, Sensory Profile, Sensory Integration Therapy

Eşik Altı Dikkat Eksikliği Hiperaktivite Bozukluğunun Dikkatsizlik Baskın Görünümünde (DEHB-I) Olan Çocuklar Duyu Bütünleme Terapisi İle Tedavi Edilmeli Mi? Bir Vaka Kontrol Çalışması

ÖZET

Amaç: Duyu bütünleme terapisi, davranışsal ve gelişimsel bozukluklar için umut verici olan önleyici terapi seçeneklerinden biridir. Bu çalışma, Dikkat eksikliği hiperaktivite bozukluğunun dikkatsizlik baskın görünümünde (DEHB-D) olan okul çağındaki çocuklar için duyu bütünleme terapisinin etkinliğine dair bir fikir vermektedir.

Gereç ve Yöntem: Tek kollu bir klinik çalışma olan bu çalışmaya, ağırlıklı olarak dikkatsizlik baskın görünümde olan 7-10 yaş arası 20 eşik altı DEHB-D tanısı olan çocuk dahil edildi. Duyu bütünleme terapisi, duyusal modülasyon ilkeleri ve müdahale stratejilerine uygun olarak hazırlanmış ve haftada iki seans olacak şekilde 12 hafta devam etmiştir. Etkinlik, Conners Öğretmen / Ebeveyn ölçekleri, Klinik Global İzlenim ölçeği, Kanada Duyusal Bütünleştirme Performans Ölçümü ve Duyusal Profil kullanılarak değerlendirildi.

Bulgular: Duyu bütünleme terapisi sonrasında, duyusal profilin işitsel işlemleme alanında "tipik" veya "daha iyi" performansa sahip hastaların oranı, önemli ölçüde artmıştır; terapi öncesinde 9 hasta (% 45); terapi sonrasında 15 hasta (% 75) (p = 0,031). Duyusal Profilin dikkatsizlik-dikkat dağınıklığı faktör puanında "tipik" veya "daha iyi" performans gösteren katılımcıların oranları, duyu bütünleme terapisinden sonra anlamlı olarak artmıştır; öncesi 6 (% 30) ve sonrası 16 (% 80) (p = 0,006).

Sonuç: Duyu bütünleme terapisi, özel gereksinime sahip olan kişilerin önemli ve amaçlı buldukları günlük yaşam aktivitelerine katılmalarını desteklemeye odaklanır. Bu çalışmada eşik altı dikkat eksikliği hiperaktivite bozukluğu olan bireylerin yaşadıkları zorluklar ele alınmış ve farklı duyusal modaliteler ile ilişkili olabilecek günlük yaşamın yönleri araştırılmıştır. Bu çocuklarda özellikle Duyu bütünleme terapisi yoluyla iyileştirilebilen bozulmuş işitsel işlemleme süreçleri gözlenmiştir. **Anahtar Kelimeler:** Dikkat Eksikliği Hiperaktivite Bozukluğu, Duyu Bütünleme Terapisi, Duyusal Profil

INTRODUCTION

Attention deficit hyperactivity disorder (ADHD) is a neurodevelopmental disorder characterized by hyperactivity, impulsivity, and inattention, which are judged excessive for the child's age or level of the overall development (1). A study of ADHD prevalence using a populationbased sample, multiple informants, and DSM-IV criteria reported that the overall prevalence of ADHD was 15.5% (2).

The symptoms are multifaceted and affect cognitive, academic, behavioural, emotional, and social functioning (3). Thus, children with ADHD may experience a number of difficulties such as academic failure, substance misuse, behaviour problems, poor peer relationships and impaired psychosocial functioning when they become adolescents or adults (4-6). ADHD can accompany difficulties in activities of daily living (ADLs), instrumental activities of daily living (IADLs), education, rest and sleep, leisure, play, and social participation (4). Similarly, children with subthreshold ADHD symptoms may have negative experimentations in daily life which are poorer academic, achievements, lower self-esteem, and poorer relationships with family members and peers (7). It is known that there is an interaction between (psychological, neurological and the child behavioural functions) and child's environment in the occurrence of these complaints (2). Previous studies indicated that sub-threshold ADHD symptoms may be formed in a part of children who are possibly more reactive to environmental risk factors (7).

Researches showed that the prevalence of sub-threshold ADHD reached to 11.7 % in the 6-12 age group children (9,11). These studies showed that sub-threshold cases were found to be more prevalent than full syndrome cases. Also, Kim et al. (9) reported that the comorbidity rate, except for anxiety disorders, was similar between full-disorder and sub-threshold ADHD and they found higher rates of internalizing problems in children with subthreshold ADHD. It was furthermore shown that in a follow-up study, the sub-threshold cases in young adulthood have a predictive importance for full syndrome disorders in later adult years (8). And this study reported that the prevalence of sub-threshold ADHD was estimated to 5.9% (8). Therefore, addressing the sub-threshold cases and applying appropriate aimed inhibition strategies are of great importance to prevent full-syndrome disorders (12).

With its focus on enabling occupation, sensory integration therapy, a nonpharmacological approach addressing activity disruptions is an important component of psychiatric treatment (13). Sensory integration therapy focuses on casecentered approaches to facilitate daily life with meaningful works (14). Sensory integration therapists are able to fulfill some needs in daily routine activities about social and motor skills, cognition, impulsivity, inattention, and hyperactivity (5). Especially family and child focused intervention programs have an improving effect on cognitive, sensorial, locomotor and play related fields (5). The antecedence of sensory integration therapy interventions is conformation to the environmental conditions, resolution of sensory integrative dysfunction, satisfaction appropriate solutions to developmental and functional problems, training of families and administration of education for ADHD (15).

There have been a few studies about the role of sensory integration therapy in ADHD (16,17). Yet, there is no study in the literature specifically investigating the effect of sensory integration therapy for sub-threshold ADHD, to the best of our knowledge. Since, sensory integration therapy is one of the promising preventive therapy options for ADHD it may have a role in the management of sub-threshold ADHD as well. The aim of this study is to provide a deeper perspective on the impact of the sensory integration therapy interventions for school-aged children with sub-threshold ADHD symptoms and to increase the awareness of the parents about their child's problem.

MATERIAL AND METHODS

The study was a single-arm clinical trial (ClinicalTrials.gov Identifier: NCT03976570) conducted at the child and adolescent psychiatry outpatient clinic of a tertiary referral hospital and involved 20 children with sub-threshold ADHD predominantly inattentive subtype (ADHD-I). Patients aged 7-10 who applied to the outpatient clinic between January 2018 and July 2018 were included in the study. The reason for selecting this age range was in terms of remaining within the same developmental period. Middle childhood is a stage when children are increasingly developing their own social, emotional and physical skills that will be needed in adolescence. Patients not adhering to regularly scheduled follow-ups were excluded from the study. Other reasons for exclusion were a personal history of any comorbid psychiatric disorder, mental retardation, learning disability, audio/visual impairment and psychiatric medicationuse. The study was approved by the Erzurum regional education and research hospital ethics committee.

Structured psychiatric meetings were conducted with the children whose parents complained of inattentive symptoms but a diagnosis of ADHD could not be confirmed with the Conner's Teacher and Parent Rating Scales (CTRS-R:S and CPRS-R:S, respectively) and the Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-V) criteria(American Psychiatric Association, 2013). The proposed DSM-V criteria for sub-threshold ADHD were used to identify patients. All of the children were evaluated with the schedule for affective disorders and Schizophrenia for schoolage children-present and lifetime version (KSADS-PL), and ADHD Rating Scale was given to teachers and parents in order to determine eligibility. The same child and adolescent psychiatrist, the lead author, administered the interviews and rated the severity of the child's illness on the Clinical Global Impression (CGI) scale at the time of assessment. Then the child and parents were referred to the sensory integration therapist for the implementation of Sensory Profile and the designation of a clientcentred therapy program for each child and parent.

The sensory integration intervention was prepared in accordance with sensory modulation principles and intervention strategies and lasted 12 weeks with two sessions per week. Every session was set to approximately 1 hour. The therapy included the interpretation of assessment results, treatment planning with parents, behavioural management of the child, environmental adaptation, classroom management, feedback session and goalsetting treatment regulating with families and children. Tactile (Brush, containers filled with beans, tactile discs, different types of fabric, ball pool, river road balance stones, shaving foam, climbing wall, ramp cushions, balls of different sizes and shapes, stones), proprioceptive (heavy suit, exercise ball, vests with pockets, ball pool, tunnel, climbing wall, cloth ball, double coordination bicycle with handle) and vestibular (trampoline, ramp cushions, balance board, swing, river road balance stones, bowl, ball pool, climbing wall, hammock) senses were studied in these children.

The effectiveness of the sensory integration therapy in the management of sub-threshold ADHD was assessed using the Conner's teacher and parent scales, the CGI form and the Canadian Sensory integration Performance Measure (COPM) before and after the intervention. Tests measuring sensory skills were administered by the same sensory integration therapist.

Instruments used:

1. The K-SADS-PL: This test is a semistructured interview form used to detect psychopathologies in children and adolescents (18). Interviews with children were conducted by a child and adolescent psychiatrist. The diagnoses were revised according to the DSM-V criteria.

2. The Sensory Profile: The Sensory Profile is a questionnaire that definesanswers to sensory events in daily life, is filled out by the parents. It is a likert scale showing how frequently the child uses that reply to certain sensory incidents (higher scores reflect higher performance). This tool scores the effects of sensory processing on a child's performance with a total of 125 items (19). The assessed sensory sections included: (1) Sensory processing, (2) Modulation, (3) Behavioral and emotional responses.

3. CPRS-R:S: This standard measure is used as a diagnostic tool of ADHD. Ithas 27 items, each item rated on a Likert scale (0=not true at all to 3=very

much true). The subscales are divided into 4 groups which are oppositional, hyperactivity, cognitive problems and ADHD group (20).

4. CTRS-R:S: This scale is mostly used to measure behavioral problems related to ADHD. There are 28 items in this scale. The subscales are divided into three groups which are Oppositional, Cognitive Problems/Inattention, and Hyperactivity (20).

5. COPM: This measure is a semi-structured interview to assign targets in the fields where the child has difficulty with self-care, creativity or play. In our study, three or four targets are chosen for each child, and after then families and children scored their performance and satisfaction scales (with a 10-point scale). Baseline and post-therapy scores were scored separately. Two or more points constitute significance (21).

6. CGI Scale: This scale is a short observation that the clinician evaluates the functioning of the patient. There are two subdivisions in which the disease assesses severity and improvement (22).

Statistical Analysis: The sample selection consisted of children with sub-threshold ADHD predominantly inattentive subtype (ADHD-I). For sample size selection, a study comparing the COPM scores to measure sensory integration performance outcomes before and after sensory integration therapy for children with ADHD was analysed (23). Median performance scores of COPM before and after intervention were 3.55 (min-max: 2.00-5.25) and 7.43 (min-max: 5.20-8.25) respectively. Treatment effect was so obvious that a minimum sample size of 2 was calculated on the basis of a hypothesis that would yield results sensitive enough to reveal a similar difference while the alpha level for rejecting the null hypothesis was set to 0.05. However, in this calculation the data was assumed to be parametric. Besides, such a treatment effect might be less evident in sub-threshold ADHD. Thus, it is decided that ensuring at least ten-fold oversampling is necessary (i.e. 20 patients). Statistical Package for the Social Sciences (IBM Corporation, Armonk, NY, USA) version 20.0 was used for the analyses. Normality was determined by Shapiro Wilk test. Descriptive statistics were expressed as follows; mean, standard deviation, and percentage. Continuous variables with normal distribution were indicated with the mean and standard deviation, and those without normal distribution with the median and interquartile range. For nonparametric conditions, Wilcoxon signed-rank test was used. Pearson correlation analysis for parametric data and Spearman correlation analysis for nonparametric data were used. For paired 2x2 table comparisons McNemar Test was used. Statistical significance limit was accepted as p < 0.05.

RESULTS

There were 20 participants who were generally low-middle-income, school-aged children. The ethnicity of all children was Caucasian. The median age of the patients was 8 (IQR: 7-10). The median age of the patients was 8 (IQR: 7-10). There were 11 male (55%) and 9 females (45%). Sociodemographic characteristics of the childrenare givenin Table 1.

Table 1. Demographic characteris	stics	ristics
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Gender, n (%)	
Male	11 (55)
Female	9 (45)
Academic performance, n (%)	
Poor	8 (40)
Moderate	8 (40)
High	4 (20)
Education of father, n (%)	
High school or lower	19 (95)
University or higher	1 (5)
Education of mother, n (%)	
High school or lower	18 (90)
University or higher	2 (10)
Intelligence score, mean \pm SD	99.8 ± 5.6
School year, n (%)	
1	6 (30)
2	6 (30)
3	2 (10)
4	2 (10)
5	4 (20)

The Sensory Profile, CTRS, CPSQ and COPM Scores of the participants were compared before and after the sensory integration therapy. Median performance scores of COPM before and after intervention were 3.55 (min-max: 2.00-5.25) and 7.43 (min-max: 5.20-8.25) respectively. The mean behaviour emotional response category score of the Sensory Profile was significantly higher after the therapy; 96 (\pm 13) before and 100 (\pm 12) after (p= 0.036). Median inattention-passivity domain score of CTS was significantly lower after the therapy; 11 (IQR: 8-13) before and 8 (IQR: 6-10) after (p=<0.01). Median inattention-passivity domain score of CPSQ was significantly lower after the therapy; 6 (IQR: 3-8) before and 5 (IQR: 3-7) after (p=0.002). Comparison of categorical domains of the Sensory Profile, CTRS, CPSQ and COPM Scores of the participants with respect to the time is displayed in Table 2.

Table 2. The Sensory Profile, CTRS, CPSQ and COPM Scores

	Week 0	Week 12	Pvalue
The Sensory Profile			
Sensory processing, median (IQR)	273 (252-292)	277 (260-285)	0.064
Modulation, mean (±SD)	126 ± 34	128 ± 17	0.293
Behavior emotional response, mean (±SD)	96 ±13	100 ±12	0.036*
Dunn total score, median (IQR)	496 (449-530)	506 (470-528)	0.062
CTRS, median (IQR)			
Inattention-passivity	11 (8-13)	8 (6-10)	0.000*
Hyperactivity index	4 (2-9)	4 (2-9)	0.603
CPSQ, median (IQR)			
Conduct problem	2 (1-5)	2 (1-5)	0.023*
Inattention-passivity	6 (3-8)	5 (3-7)	0.002*
Hyperactivity index	4 (2-9)	4 (2-7)	0.153
Oppositional index	2 (2-4)	2 (1-4)	0.161
СОРМ			
Performance, mean (±SD)	3 ±1.37	5.5±1.96	0.00*
Satisfaction, median (IQR)	3 (2-4)	6 (4-8)	0.00*

CTRS: Conner's Teachers Rating Scale, CPSQ: Conner's Parents' Symptom Questionnaire, COPM: Canadian Sensory integration Performance Measure, * stands for p <0.05.

Individual scores of specific domains in the Sensory Profile were analysed after categorization with respect to the normative data, and only the rate of patients with typical or better performance in auditory processing domain were found significantly increased after sensory integration therapy; 9 patients (45%) before and 15 patients after (75%) (p=0.031). The rates of participants withtypical or better performance in all domains before and after the sensory integration therapy were displayed in Table 3. The rates of participants with typical or better performance in inattentiondistractibility factor score of the Sensory Profile were found significantly increased after sensory integration therapy; 6 before (30%) and 16 after (80%) (p=0.006).

The psychiatrist's ratings on the CGI scale indicated that symptoms of sub-threshold ADHD dramatically improved in 20% of the patients, minimally improved in 70% and there was no change from baseline after therapy in 10%. None of the participants showed any deterioration during the course of the program.

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Table 3. Typical or better performance rate in the section summary outcome of the Sensory Prof	ile, n
(%)	

Item Categories	Before therapy	After therapy	p value
Sensory processing			
A. Auditory processing	9 (45)	15 (75)	0.031*
B. Visual Processing	15 (75)	17 (85)	0.500
C. Vestibular Processing	8 (40)	12 (60)	0.219
D. Touch Processing	15 (75)	15 (75)	1.000
E. Multi-sensory Processing	13 (65)	17 (85)	0.125
F. Oral sensory processing	15 (75)	16 (80)	1.000
Modulation			
G. Sensory processing related to Endurance/Tone	5 (25)	7 (35)	0.500
H. Modulation Related to Body Position & Movement	5 (25)	4 (20)	1.000
I. Modulation of Movement affecting activity Level	15 (75)	16 (80)	1.000
J. Modulation of Sensory Input affecting Emotional Responses	12 (60)	13 (65)	1.000
K. Modulation of Visual Input Affecting Emotional Responses and Activity Level	15 (75)	17 (85)	0.500
Behavioral and emotional responses			
L. Emotional/Social Responses	13 (65)	14 (70)	1.000
M. Behavioral outcomes of Sensory Processing	6 (30)	11 (55)	0.063
N. Items indicating Thresholds for Response	17 (85)	17 (85)	1.000

DISCUSSION

As seen in previous studies, it was found that children with ADHD diagnosis had lower sensory profile scores than the control groups (15,24,25). However, no study has been found on children diagnosed with sub-threshold ADHD-I. In our study, a significant improvement was observed especially in the auditory area in patients with subthreshold ADHD-I symptoms. This condition may suggest that the first degraded area was the auditory area. Perhaps when this problem in the auditory areas is detected early, precautions that can be effective in treatment can be taken and full syndromes can be prevented with the necessary sensory integration treatment. This remark served as an effective idea to identify difficulties of children with sub-threshold ADHD and it was certainly a useful starting point for formulating potential improvement plans thereto. The probable difficulties experienced by children with subthreshold ADHD-I are addressed and aspects of daily life are explored while swiping through different sensory modalities. As a result, an intriguing observation was made. That is the importance of auditory processing in this population.

The study brought new insights into the plasticity phenomenon of the human brain.Plasticity is described as the capability to modify the structure

and/or function of the nervous system. It is related to the sensory experience in the auditory cortex (26). Although, sub-threshold ADHD-I is not a complete sensory deprivation due to hearing loss this disability might share some common pathophysiological mechanisms.

Previous studies have generally shown that sensory integration therapy increased improvements in both goals and motor performance in ADHD patients (27). And this study shows it works for sub-threshold ADHD-I children as well. According to CPRS-R:S ratings on conduct problem subscales, problematic behaviours may also be ameliorated. This situation can be explained by the fact that the child is more likely to cooperate as a result of the skills gained after this therapy. So, the stress experienced by families the may also decrease.Inattention problems such as, having a short attention span and being easily distracted, making careless mistakes, appearing forgetful or losing things; all might be ameliorated with a simple sensory integration intervention. Being unable to stick to tasks which are tedious, appearing to be unable to carry out instructions, constantly changing activity, having difficulty organising tasks and following with parental directions may all be improved with sensory integration therapy. During the first interview, the child's psychological and

sensory tests were performed. Descriptive tests were conducted on the child's performance. Information about the child's performance was transferred to the family. The family was asked to transfer the information to the teacher. This is especially important in determining the common goals with families and teachers. The necessity of this situation for a better treatment is emphasized in some studies (28,29).

A target treatment program was set for each child according to COPM scores and there was a significant improvement in both of the performance and the satisfaction scores. After the therapy sessions, only about 10% of the children were rated by psychiatrist as "no change from baseline" compared with their status at baseline and there was no worsening child. Parents and teachers reported an improvement after the sensory integration therapy sessions on Conner's inattention-passivity subscales. Likewise, parent's ratings reflected significant changes on Conner's conduct problem subscale. However, both of CPRS-R:S and CTRS-R:S ratings on hyperactivity index scales reflected no significant changes after the therapy. The inattention-passivity subscales arevery important not only for children with sub-threshold ADHD children but also for their parents.

Yet, this study has several limitations. First the sample size was small. Second, investigators and participants were not blinded to the therapy conditions. Thus, the ratings were subject to observer bias. Despite these limitations, the results of this study suggested that the sensory integration therapy on sub-threshold ADHD could be successfully applicable especially inattentionpassivity symptoms.

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